Diagnostic uncertainty in youth with chronic pain and their parents

Alexandra Neville[1], Abbie Jordan[2], Jaimie Beveridge[1], Tamar Pincus[3], Melanie Noel[1]

[1] Department of Psychology, University of Calgary and Alberta Children's Hospital Research Institute, Calgary, AB, Canada

[2] Department of Psychology and Centre for Pain Research, University of Bath, United Kingdom

[3] Department of Psychology, Royal Holloway, University of London, United Kingdom

Corresponding Author:
Alexandra Neville
Email: alexandra.neville@ucalgary.ca
Department of Psychology, University of Calgary and Alberta Children's Hospital Research Institute
2500 University Drive NW
Calgary, AB, T2N 1N4, Canada

Disclosures: This work was supported by funding awarded to Noel by the Vi Riddell Pediatric Pain Initiative, the Alberta Children's Hospital Foundation and Research Institute (Shaikh Award), and funding from the Canadian Institutes of Health Research Strategy for Patient Oriented Research 'Chronic Pain Network'. The authors have no conflicts of interest to declare.

Abstract

Diagnostic uncertainty, the perception of a lack of or incorrect label to explain symptoms, has been reported by parents of youth with chronic pain. This study was the first to examine diagnostic uncertainty in both youth with chronic pain and their parents using qualitative methodology. Individual, face-to-face, semi-structured interviews were conducted with twenty youth with chronic pain recruited from a pediatric chronic pain program. Independent interviews were also conducted with one of their parents. Interviews explored participants' memories and perceptions around diagnosis. In-depth thematic analysis revealed four themes: (1) The Function of a Diagnosis. Parents and youth struggled with the meaning of the diagnosis, needed further explanation for the pain, and perceived the 'right' diagnosis (i.e., one that fit with their beliefs) as justification for the pain. (2) Haunted by Something Missing. Negative test results did not provide relief or counter the belief that something serious could have been missed by clinicians. (3) The Search for an Alternative Diagnosis. A search persisted for the 'right' diagnosis, particularly when a non-pharmacological treatment plan was provided. (4) Mistrust in the Medical System. Clinician communication and perceptions of clinicians' uncertainty impacted parent and youth 'buy in' to the diagnosis. Findings suggest that many youth with chronic pain and their parents experience diagnostic uncertainty, which is integrally tied to their past experiences with the medical system. Greater understanding of diagnostic uncertainty may help tailor how clinicians deliver diagnoses to achieve 'buy in', increase understanding of pain and diagnosis, and improve treatment response.

Perspective: A major challenge that youth with chronic pain and their parents face is understanding the cause of the pain. Youth with chronic pain and their parents experience uncertainty about their diagnosis, which may be linked to their 'buy in' and treatment response.

Diagnostic uncertainty in youth with chronic pain and their parents

Pediatric chronic pain, affecting 11-38% of youth,[17] can have a profound impact on daily functioning, and is associated with frequent school absences, sleep disturbance, emotional distress, and reduced participation in peer activities.[29] The impact of pediatric chronic pain extends to the broader family. For example, parenting a child with chronic pain is associated with anxiety, depressive symptoms, and parenting stress.[5] A major challenge that children with chronic pain and their parents face is understanding the cause of the pain. Indeed, the nature of a chronic pain diagnosis is inherently complex, challenging to explain and understand, and fraught with uncertainty. The time spent investigating the child's pain in search of a diagnosis and cure has been described as a "diagnostic vacuum" and may be critical to parents' and children's pain conceptualization and treatment.[6] Among adults with chronic pain, diagnostic uncertainty, recently defined as a subjective perception of an inability to provide an accurate explanation of the patient's health problem,[1] is associated with higher levels of anxiety, depression, pain intensity,[33] and disability.[12] However, an understanding of how diagnostic uncertainty is experienced by youth with chronic pain and their parents is unclear.

Diagnostic uncertainty has been alluded to in the pediatric chronic pain literature and a recent review has raised it as a critical issue for further investigation in this population.[30] In a study of 53 children with chronic pain, the majority of children perceived the explanations provided by physicians for their pain as false.[22] Two studies have explored diagnostic uncertainty among parents of youth with chronic pain. In a qualitative study, Jordan and colleagues[16] described parents' struggle to accept a lack of explanation for their child's pain and an ongoing, unrelenting search for a diagnosis that would validate the pain, even after a diagnosis of chronic pain had been provided. Uncertainty with regards to diagnosis was tied to parental guilt,

helplessness and distress.[16] Parents also described a sense of being "in limbo," a state of uncertainty in which they could not move forward in daily life and felt unsure about the future.[16] In a later observational study, nearly 40% of parents reported an unresolved diagnostic orientation towards their child's chronic pain (i.e., not accepting or 'buying in' to the diagnosis).[26] This unresolved orientation was found to be a critical aspect of a distress narrative among parents of children with chronic pain, which in turn was associated with increased helplessness.[26]

Given the lack of research on diagnostic uncertainty with regard to pediatric chronic pain from the perspectives of youth and their parents and its potential impact on treatment outcomes, this study employed a qualitative methodology to investigate how diagnostic uncertainty is experienced by *both* youth with chronic pain and their parents. A dyadic qualitative approach to examine this phenomenon offers an opportunity to elucidate the complex experience of diagnostic uncertainty from the unique perspectives of youth and parents that may not be conveyed through questionnaire data.

Method

Participants

Twenty youth with chronic pain (15 female, M_{age} = 14.6 years, range = 10-18 years) and one of their parents (17 mothers) each participated in separate face-to-face interviews with a researcher. Youth and parents were recruited from an interdisciplinary, tertiary-level pediatric chronic pain program in Western Canada. This team was led by an Anesthesiologist and included clinicians from Anesthesiology, Psychology, Nursing, Physiotherapy, and Family Therapy. At the time of the interview, all participants had been at least initially seen in the pain clinic. *Y*outh were eligible to participate if they were 10-18 years old and reported a pain problem that had

been present for at least three months (in accordance with the current definition of chronic pain endorsed by the International Association for the Study of Pain).[23] Exclusion criteria for youth included a diagnosis of a serious medical condition (i.e., juvenile arthritis or cancer), neurodevelopmental disorder (e.g., intellectual disability, autism spectrum disorder), and/or a serious mental health disorder (e.g., schizophrenia, psychotic disorder). Parents were eligible to participate if they were the legal guardian of the youth.

Youth were referred to and/or attending the headache (n=8) or complex pain (n=12) clinic of a tertiary-level pediatric chronic pain program. Of youth who reported on their pain duration (n=16), the average pain duration was 3.67 years (SD = 3.32 years, range = 0.5–11 years). Youth (n=20) reported an average pain intensity of 5.95 out of 10 (SD = 1.55) in the past week. During the interview, parents and youth were asked whether or not a diagnosis for the youth's pain had been provided by a clinician. Sixteen parent-youth dyads were in agreement in terms of the diagnosis that the youth had received: complex regional pain syndrome (n=6), headaches/migraines (n=2), migraines and stomach migraines (n=1), migraines and constipation (n=1), chronic pain (n=2), nephroptosis (n=1), costochondritis (n=1), no diagnosis (n=2). However, among 4 parent-youth dyads, either the parent or youth reported a diagnosis of migraines, while the other member of the dyad reported that no diagnosis had been provided (parent reported migraine diagnosis n=1, youth reported migraine diagnosis n=3). Information with regards to these diagnoses were reported by families to have been provided by a variety of clinicians/teams, including the pain clinic, emergency physicians, neurologists, general practitioners, nurses, psychologists, and physiotherapists. Additional sociodemographic information (e.g., ethnicity, household annual income) are presented in Table 1. The current sample size is consistent with previous studies using thematic analysis.[35, 36] In accordance with

the views of Braun and Clarke, no attempt was made to achieve saturation within the data as this is an unhelpful concept.[28] Instead, a focus was placed on establishing a transparent process around how the data was collected, as research transparency is perceived as a marker of quality.[37]

Procedure

All study procedures were approved by the institutional health research ethics board (REB). Parents were asked by a member of the pain clinic staff for permission to be contacted by the research team. A member of the research team contacted permitting parents over the telephone or via email with information about the study. A consent call was performed with parents and youth to determine eligibility and answer any questions about the study. Interested parents and youth were then emailed separate online consent and/or assent (for youth aged 10-13) forms via Research Electronic Data Capture (REDCap), a secure online data collection tool,[14] to complete prior to the interview. On the day of the interview, hardcopy written, informed consent/assent was also obtained from youth and parents. Youth and parents were informed that interviews would be audio-recorded and provided permission for quotations from the interviews to be published for academic purposes.

Interviews with youth and parents were conducted separately in a private testing room at the hospital where the chronic pain program is located. Interviews were conducted in-person by members of the research team (the PI [clinical psychologist] or clinical psychology graduate students with previous training and experience interviewing youth in clinical and research settings). The same researcher interviewed both the youth and parent of a dyad, with the youth being interviewed first and the parent immediately afterwards. Interview duration ranged between 10-50 minutes (mean duration youth interviews = 19 minutes, mean duration parent

interviews = 27 minutes). Interviews were audio-recorded and transcribed verbatim by members of the research team. All identifying information was removed from the transcripts to ensure the anonymity of participants and related individuals. Youth and parents both received a $25 CAD gift card after the visit to thank them for their time.

Interview Schedule

Researchers with expertise in qualitative research methods, diagnostic uncertainty, and pediatric chronic pain (MN, AJ) developed the interview schedule (see Table 2). The interview schedule was designed to gain insight from the youth and parent about their 'pain journey' (i.e., when the pain started, what living with pain has been like, perceptions regarding future outcomes with regard to pain and life). One question in the interview schedule (item #4 in Table 2) focused specifically on the youth's and parent's experience of obtaining a diagnosis for the pain. Youth interviews focused on the experience of living with chronic pain whereas parent interviews focused on the experience of parenting a youth with chronic pain.

The interview schedule was semi-structured and comprised open-ended questions in order to allow interviewers to follow-up and expand on topics of interest as they occurred during the interviews.[21] Specific prompts were included in the interview schedule to encourage elaboration on topics of interest (e.g., 'Tell me more about that?').

Data Analysis

Data were analyzed using reflexive thematic analysis, in accordance with Braun and Clarke's[2] six-phase thematic analysis guide. Specifically, familiarization with the data was achieved through repeated and active reading of the interview transcripts. Through iterative familiarization with the data, codes were generated for data items related to receiving a diagnosis and then collated to identify potential themes. Initial analyses to generate codes and identify

potential themes were conducted by co-author AN. AN first reviewed all youth interviews, followed by all parent interviews. Themes were then compared across each parent-youth dyad to identify patterns that were common and differed within dyads. Throughout this process, frequent debriefing sessions were conducted with AN, JB, and MN, to discuss the development and interpretation of themes. Co-authors AJ and TP frequently reviewed the ongoing analyses and debriefed with authors AN, JB, and MN (e.g., discussion of alternative interpretations, need for further analysis).[34] Potential themes were iteratively reviewed and refined to determine the final themes, and concise definitions for each theme were generated. All co-authors were involved in, and agreed upon, the analyses and interpretations, providing credibility and trustworthiness in the data and analytic interpretations.[8, 24] Lastly, the data were analyzed within the identified themes, and a report was written. In keeping with the principles of an inductive thematic analysis approach, we did not count frequencies of themes. Indeed, the importance or meaningfulness of a theme does not necessarily equate to frequency or quantifiable measures.[2]

Data analysis was completed using QSR International's NVivo software, a computer-assisted qualitative data analysis package. Attention was paid to include quotations from various participating youth and parents for representation of all perspectives within each theme. The consolidated criteria for reporting qualitative research (COREQ) checklist was followed to ensure a rigorous and thorough approach to establishing quality and reporting of this qualitative study.[39]

Results

Four themes were generated from thematic analysis of the data: The Function of a Diagnosis, Haunted by Something Missing, The Search for an Alternative Diagnosis, and Mistrust in the Medical System. These themes are presented below with supporting verbatim

quotes from youth and parents. Pseudonyms are used below for each participant to protect their confidentiality. In addition, we use the term 'clinicians' below to refer to all types of clinicians who may provide information to families about a chronic pain diagnosis (e.g., anesthesiologists, neurologists, rheumatologists, general practitioners, nurses, psychologists), recognizing variability within this sample and across contexts (e.g., pain teams, internationally).

1. The Function of a Diagnosis

When provided with a diagnosis for their chronic pain from a clinician, many youth and their parents struggled with the meaning of their diagnosis. For many, a label was not enough and the diagnosis provided little clarity in terms of understanding their pain. Parents and youth alike expressed needing further explanation beyond a label for their pain.

> *"Uh, they said chronic regional pain syndrome. Mm, a whole bunch of words that really tell us nothing." (Mother of Lauren)*

When youth and parents reported understanding the label provided for their pain, further explanation about the etiology and causal mechanisms underlying the diagnosis was still needed.

> *"I've always had anxiety surrounding my pain, cause I'm like 'I don't know why this is happening.' And I think… a lot of my anxiety is like when I don't know why something's happening I'm like 'I need to know why' and I don't know why I have migraines… I know what a migraine is but I don't know why I have them." (Isabelle, age 17)*

A diagnosis of 'chronic pain' itself seemed to elicit uncertainty and was perceived by parents as a diagnosis given either in the absence of a true understanding of the cause of the pain or as a "catch all" or "default" diagnosis, which was unsatisfactory for parents.

> *"Um, and it was like kinda c- called chronic migraines and we have zero history of that in our family right… So the whole thing was just perplexing. But I understand like I think it's probably a default diagnosis." (Mother of James)*

Discrepancies were found among some parent-youth dyads in how they made sense of or felt about the diagnosis. For example, while Cecilia seemed to express that the diagnosis carried little meaning and only fit "to a certain extent", her mother reported experiencing relief in having a diagnosis. For this parent, a new explanation that had not previously been provided and that no longer focused solely on common explanations for the pain (e.g. nerve pain) was reassuring and validating.

> *"It's just a diagnosis to me. It's just a word… it's just a word that they think is what I have… It's just a label that they put the pain I have under." (Cecilia, age 15)*

> *"It was just a relief to have someone that actually gave you a possible answer for what had been going on all this time as opposed to, you know, 'well it's just pain, you just have to wait for this, here's some tablets off you go'… It just seemed, it was like a completely different answer as opposed to just nerves or anything else, it was just a term I hadn't heard before. But it was, I, I don't know it just, it just seemed something that fitted I suppose… You know, someone doesn't throw out a random term just for the hell of it." (Mother of Cecilia)*

In the absence of a diagnosis from a clinician, some youth's pain was disbelieved by others, including parents, peers, and clinicians. Parents and youth described a diagnosis as external validation that would justify the pain. Being disbelieved by parents was particularly salient perhaps because of the widely held belief that a parent should unconditionally believe their child. For example, the mother of Olivia described receiving a diagnosis as a "game changer" in that it allowed the father to finally believe that his daughter's pain was real.

> *"But all of a sudden there was a diagnosis and all of sudden Dad's on board and he's there. And he's supportive and which is what she needs. So the diagnosis was a bit of game changer for him. For him like oh she isn't just faking it, she isn't just a being dramatic, she isn't being a girl." (Mother of Olivia)*

Emily's mother explained how, to Emily, a diagnosis meant a physical cause or explanation that would 'prove' the validity of the pain to clinicians. For Emily, a 'chronic pain' diagnosis conveyed that her pain wasn't real and wasn't believed. Emily explained that a physical abnormality in her wrist to explain her chronic pain problem was more reassuring than a diagnosis of complex regional pain syndrome.

> *"I think she has distrust in the system and they still don't get her… even our GP, [emergency doctor], nobody gets her or trusts that it's real pain. And I think her definition of real pain may be a little bit, like she needs a physical diagnosis to justify her pain." (Mother of Emily)*

> *"They just told me I have chronic pain and I was like a bit upset, I was like excited that there was a diagnosis, but I was a bit upset because... I thought that chronic pain was like your brain... I was upset that like people didn't think I had real pain. So, I got really upset… then they told me there is still something. My wrist clicks and stuff, so there is still something in my wrist that is happening." (Emily, age 11)*

For some, uncertainty extended beyond the diagnosis to the treatment for the pain. In addition to the diagnosis not providing clarity, a non-pharmacological treatment plan (e.g. physiotherapy or psychology) fuelled further confusion, ambiguity, and uncertainty and was seen as less valid than a pharmacological intervention.

> *"It's not this, it's not this, it's not this. They're not telling you what it is. And even when they do tell you what it is, then you know, well the main therapy is physio which is great 'cause it's something you can do but then as soon as you hear that, you know it's something that's going to take time and then you don't really know. This is a very ambiguous thing from what I can tell to be diagnosed with." (Mother of Jada)*

> *"And my doctors have said like maybe you have endometriosis. And I'm like well, like what the hell is that. Like how does that help me? You can't give me medication for it." (Jada, age 17)*

Relief was anticipated if the 'right' diagnosis (i.e., one that fit with the parent and youth's perception of the cause of the pain and anticipated management) was discovered. Furthermore,

the ‘right’ diagnosis was perceived as being one that required a pharmacological solution and ‘silver bullet cure’, which would abolish the pain entirely.

> *“I think if I were to finally get the right diagnosis it would be like a huge relief. Because then I would know if I can be taking medication for it to help it or if I could be doing something to help it or anything. Like then I can just make it stop. I think that’s the best part about it. Like I can make it stop.” (Kristen, age 17)*

When treatment for a diagnosis did not lead to improvement in pain and functioning, some parents questioned their child’s pain, and believed that recovery was needed to provide evidence that the diagnosis was accurate and the pain was real.

> *“So we’re like okay, well if it’s an inflamed joint, then the inflammation pills should’ve at least eased it down. But they did actually nothing. So that’s when we were like, you know, well maybe it is in her head, maybe there is something that she’s associating this with and uh, not getting enough sleep and being overdramatic and emotional over something that’s you know, making a mountain out of a molehill kind of thing.” (Father of Lilly)*

2. Haunted by Something Missing

Uncertainty about the youth’s diagnosis fuelled worry that something could have been missed and, further, that what was being missed was serious and sinister.

> *“It makes me kinda worried to what it might be if it could be something really severe. But it’s probably not. But still that little inkling… if I have something in my brain that might not be good. Something that I might have to live with forever. Not knowing what it might be that’s also in that we know that ‘something’s bad we just can’t tell you what’ kind of feeling…” (Nicole, age 14)*

Thoughts and beliefs that something could be missed were intricately tied to previous experiences with clinicians and the medical system. Long periods waiting in a state of uncertainty seemed to feed a lack of acceptance of a diagnosis when it was finally provided. For some, fear and health anxiety developed while searching and waiting. In this way, delays in

receiving a diagnosis and repeated failed explanations for the pain further fuelled diagnostic uncertainty.

> *"…like because it took so long to figure out what was wrong with me… it enabled me to like, with my anxiety and stuff, it enabled me to kind of like just create all these different like serious problems with me because like they, they missed like if they can miss something like this [recent diagnosis], what else are they missing, kind of thing? Um so like I just have a general fear of cancer. I always think I have cancer at some point in time so. That's like my first question going into a doctor. Like, 'Do I have cancer?' 'Doesn't sound like it.' 'Okay, cool…' 'Cause like with um this endometriosis, just kind of the symptoms I have been experiencing as of late are very um, synonymous with ovarian cancer." (Olivia, age 18)*

When pain did not take the course that was perceived as promised by clinicians, uncertainty developed for Lilly that something had been missed. This bred frustration when time, even years, passed with little improvement in pain symptoms. In the midst of longing for a cure, youth doubted whether clinicians had done everything they could when the pain began and they wondered if something else could have been done if searched for earlier.

> *"I still feel slightly as if they're missing something, as if something else is wrong because they said it's going to rule itself out but they've been saying that for two years and it hasn't. So I think it's just the frustration like, I just want to be fine. I want to be able to run without feeling like I can't breathe and fainting. I want to be able to leave my house and not be in pain constantly… So I think it was just the sake of- I was like did they catch everything they could've?" (Lilly, age 15)*

For others, the fear that they could be missing the cause of the pain was fuelled by guilt and by consequences of past experiences when something *had* been missed. Although intricately tied to mistrust in the medical system, James' mother shouldered the guilt of having missed a diagnosis earlier in his childhood because she had accepted an inaccurate clinical diagnosis and had not advocated effectively for her child at that time. This past experience drove current worries that something sinister was being missed in explaining her child's chronic pain condition, a decade

later. For some, when explanations from clinicians were perceived as inadequate or inaccurate, the assumed responsibility of finding a resolution for the pain shifted from clinicians to the parents themselves.

> *"And what if we're missing it because we missed his [reflux]- this is where it goes back to me with the guilt right- We missed that reflux that caused the damage to the kidneys so so, I'm always now really worried, like what if we're missing something that's causing this and is gonna have long term effects. 'Cause I feel like that we missed it the first time. Right- like so that's what probably my biggest worry is." (Mother of James)*

In the face of negative test results, parents and youth struggled to make sense of a chronic pain diagnosis. Test results seemed to have ruled out all other possibilities, and yet this did not provide definitive proof of, or a cause for, the pain. Furthermore, living with chronic pain did not fit with their expectations of what a normal childhood should be.

> *"I'd go see doctors but they'd just kind of um test my blood, and I'd get CAT scans and they'd say like everything was fine. It was strange, 'cause we knew everything wasn't fine. 'Cause it's not normal to get so many headaches." (Julia, age 16)*

Parents seemed to oscillate between rationalizing that all tests had been done and feeling that there must still be a physical cause for the pain that was yet to be found. For many, negative test results did not provide relief and raised more questions. Parents and youth wanted tests to 'prove' a 'physical' diagnosis, especially one that was curable, rather than tests which continued to exclude diagnoses.

> *"I know she hasn't got any kind of like actual physical structural problems because they did an MRI last summer, but I just think it has to be some kind of, either, you know, the wonders of the world of migraines that people don't understand or stress, like a bunch of little triggers… despite my lack of uh belief in catastrophic thinking, uh if you are, if somebody's fainting 10 times a day there must be some kind of physical trigger, you'd think. There needs to be a physical trigger." (Father of Nicole)*

Parents expressed tension between the risks involved in continuing to search for a cause of the pain and the need to end the search despite never definitively knowing whether something had been missed. Parents conveyed lingering questions about how long to continue testing, whether or not tests were necessary, and when the number of tests would be enough to stop the search. This was made even more difficult as parents were making decisions about their child and not themselves.

> *"I just feel like probably the biggest thing is… from a pain point of view, I feel like I really hope we're not missing something that's causing this. And yet you don't want to do a million tests, like there's risks to MRI, there's risks to CT scans, right like, so you don't want to, you don't want to be… also, you want to be sure you're being diligent but you also want to be sure you're not doing things unnecessarily so it's a very fine balance." (Mother of James)*

3. The Search for an Alternative Diagnosis

Even after youth received a diagnosis and treatment, some parents and youth continued to search for an alternative diagnosis for the pain. When clinicians were not perceived as fulfilling their role in providing the 'right' diagnosis, this led to frustration and a pursuit for another clinician who would provide an acceptable answer, reassurance, and alternative explanations. These alternative clinicians were perceived as offering hope and promise for achieving the ultimate goal of abolishing the pain.

> *"… I had done some research and had found there is a [doctor 1] here in [City] is a specialist in endometriosis so I had asked [doctor 2] if we could get a referral... I want [Olivia] to see [doctor 1] because [she] wouldn't be satisfied until the person who is a specialist in that field tells her." (Mother of Olivia)*

Even parents and youth who perceived the medical system as being supportive reported searching for another explanation for the pain. While *both* parents and youth were in pursuit of an elusive "satisfied" result, which ultimately was a *fix* for the pain, parents took on this search as part of

their parental role. In this way, finding a resolution for the pain was part of being successful in their role as a parent.

> *"You know I've said to my husband you know, 'do you think like we should take him like, maybe we should get a consult elsewhere'… As a mom I feel like we shouldn't be stopping until we can fix this headache is what I feel like." (Mother of James)*

In some cases, the youth's need for a diagnosis, a reason, and closure for the pain was so strong that it was worth undergoing invasive surgical procedures, even when the parent was unsure it would help. As Emily's mother explains below, undergoing surgery brought hopeful expectations of a final validation for her daughter that would end her search for a cause of the pain. However, for many families, a final validation was seemingly just beyond grasp, tenuous, and intangible.

> *"Well I truly hope that when she has the [surgical procedure], she'll know that everything has been done, that can possibly be done. I don't want her to have a surgical procedure. I wish she could just- but I think for her to move forward. That's her final validation." (Mother of Emily)*

Others expressed a shift from an arduous search for the cause of the pain to a focus on function. However, within dyads, this shift did not necessarily occur in unison.

> *"I think, I think early on I was always trying to find "it", you know? If we just figure this out you know, then happy pain-free ever after. Like, "the cause" and, therefore, "the solution…" And, where I'm at now… I think it's all about function and making the best of what it is, you know?" (Mother of Julia)*

> *"I'm still not really sure where I stand with [the diagnosis] 'cause it's just very confusing like different doctors saying different things." (Julia, age 16)*

4. Mistrust in the Medical System

Uncertainty and fear that something could be missing was met with varied emotions. When individuals felt that diagnoses provided by clinicians were lacking credibility, individuals perceived a lack of voice and sense of not being heard. Beyond worry about a lack of, or unsatisfactory, diagnosis, youth expressed frustration with communication with their clinicians. A disconnect between patient and clinician agendas was sensed by youth, creating a deeper divide between clinicians and youth. This ultimately impacted their trust in clinicians and the medical system as a whole.

> *"I don't want to tell people how to do their job but I felt that nobody was doing their job for me. Like they weren't paying enough attention so I was like, 'This is what it is.' And they're like, 'No it's not.' And I'm like, 'But you just think I'm pregnant so—' 'Cause every time I went in she gave me a pregnancy test which was super frustrating because I wasn't even sexually active then… But every time I went in, it was pregnancy test and I'm like, 'Last week I wasn't pregnant, this week I'm not pregnant, let's like try something else.'" (Olivia, age 18)*

Parents also noted the divide between their children and clinicians. For Olivia and her mother, a credible plan to receive the 'right' diagnosis was celebrated. The emotional intensity was about more than the relief of having a plan, it was about being listened to and validated. Importantly, Olivia's mother anticipated that even if a negative result was found (i.e., "it's clean") the results of this procedure would lead to a subsequent plan.

> *"Again, we came out of there crying because [Olivia] was like oh my God she's awesome she heard me. That's exactly what she wanted someone to go in there and say okay if it's cleans it's clean what are we going to do about it, if it's not clean okay we're fixing it. A plan, and she was heard. I can't tell you it was like [sigh], like I'm just the adrenaline right now is just cause it's been such a long road." (Mother of Olivia)*

An explanation that didn't fully 'fit' left youth feeling dismissed, unworthy of clinicians' time, and that they had not received proper care.

"I do believe them that hormones are playing into it but it's not just hormones. It feels like they're just trying to find a way so that way they can see their other patients that have been waiting. They're just like 'it's hormones, goodbye.' I think that they are saying that for a reason but I don't like it because it makes me feel like I'm not worth the extra effort." (Nicole, age 14)

Parents and youth also perceived clinicians' own uncertainty or lack of confidence in the diagnosis. Stephanie's mother particularly expressed a lack of confidence in the amount of specialty training that the clinician providing the diagnosis had received.

"Initially I was not super accepting of [the CRPS diagnosis]. It seemed like it was 'kind of but we're not sure so', um was the feeling... And some of that you get because you end up with residents who end up with, who are, who are gracious enough to say, "We don't know." Right? So they go get an emerg doc and I've had enough experience with some of these guys and they're not specialists" (Mother of Stephanie)

Clinician communication affected not only patient 'buy in' to the diagnosis but also patient confidence in the clinicians' ability to help or provide an accurate diagnosis.

"I didn't really know what it was. Like I had never really heard of it before and he was like um, and the doctor, he was like 'You're the first person that I've ever diagnosed with this.'... So um I was just like this doesn't really make any sense. ...But um, yeah, I was just, I didn't really know what to think of it. Like, I don't know, I was just kinda confused." (Jada, age 15)

Perceived credibility of the treatment team was critically important for some families in reducing their diagnostic uncertainty. Jada reported a better understanding of the diagnosis after meeting with an interdisciplinary pain team who "knew more about [the diagnosis]". Whereas Jada expressed acceptance of the diagnosis, Catherine on the other hand, expressed continued doubts in clinician's understanding of her condition. For her, she perceived that clinicians were uncertain in the diagnosis themselves.

"I think they're missing stuff. Or they don't know what it is and they're just trying to diagnose it." (Catherine, age 11)

Even when trust in clinicians was expressed, an air of uncertainty lingered. While expressing a sense of hope that a solution would be found, unanswered questions undermined parents' ability to fully trust their child's clinicians.

"I mean I trust the radiologist has some ideas on what it is– I guess… I have that little inkling of, 'okay well, why is his bloodwork off too?' Like, 'could it be something more?' But I trust that they're gonna figure… I trust they're gonna figure it out…" (Mother of Samantha)

Discussion

This is the first study to explore the experience of diagnostic uncertainty in youth with pediatric chronic pain and their parents. In-depth thematic analysis of interview data with youth and parents resulted in the generation of four themes, which illustrate the complex nature of a chronic pain diagnosis and the experience of diagnostic uncertainty. Across the themes, which were labelled, The Function of a Diagnosis, Haunted by Something Missing, The Search for an Alternative Diagnosis, and Mistrust in the Medical System, parents' and youth's struggle to accept and understand a chronic pain diagnosis was met with frustration, disempowerment, feeling unheard and alone, and mistrust in the medical system.

Parents and youth expressed a lack of clear understanding of the diagnosis provided. For many, a diagnosis of chronic pain was not a satisfying result of medical investigations and parents and youth were left wanting a biological/physical explanation for the pain. For some youth, a diagnosis of 'chronic pain' conveyed that clinicians did not understand the pain or did not perceive the pain as being 'real.' A diagnosis from a clinician that 'fit' was a 'game changer' and was critical for others, particularly parents, to 'buy in' to the authenticity of the pain.

Without a diagnosis that was perceived as credible, however, youth felt invalidated, which is commonly reported among patients with chronic pain.[4,18] In a qualitative study of children with medically unexplained symptoms, children and their parents expressed struggling to accept a lack of a credible diagnosis and needing an explanation for their symptoms.[25] Moreover, most children reported not feeling understood, believed, or listened to by physicians.[25] Although validation has not been shown to lead to reductions in pain, invalidation during a pain tolerance task with healthy adults has been linked to a decrease in positive affect and increased worry.[19] Future research is needed to examine the impact of validation on other components of pain (e.g., disability) and in chronic pain populations.[7] Indeed, validation was a core objective in parents' and youth's search for a diagnosis. Validation of pain (i.e., communicating that pain-related thoughts and feelings are understandable and legitimate) may impact patients' emotional state (i.e., increase/maintain positive affect) which could reduce pain[7] and possibly increase orientation to and acceptance of a chronic pain diagnosis.

Uncertainty extended beyond diagnosis to treatment. Parents and youth expressed holding beliefs that the 'right' diagnosis would lead to a pharmacological treatment, which would 'fix' the pain. This same belief was found in reverse throughout parent and youth interviews, such that a reduction in, or eradication of, pain was seen as proof that the 'right' diagnosis had been found. The objective of receiving a diagnosis that would lead to a treatment that would make the child pain-free, has been previously reported by parents of children with functional abdominal pain.[3] This belief that a 'right' diagnosis would lead to a cure for the pain demonstrates a desire that is incompatible with current chronic pain treatments, which prioritizes improving function, often with the expectation/hope that this will lead to improvements in pain.[10, 20]

Parents and youth expressed uncertainty and concerns that a serious health condition (e.g., cancer) could have been missed, which fuelled an unrelenting search for the 'right' diagnosis and cause of the pain. For many, negative test results did not provide relief but rather fuelled diagnostic uncertainty while parents and youth attempted to make sense of the pain in the absence of objective evidence. Despite a widely held belief by clinicians that additional testing will reassure patients,[40] there is limited evidence that diagnostic testing reassures patients.[41] Among adults, informational reassurance (e.g., explanations about physical causes for pain) has been shown to be more reassuring than emotional reassurance (e.g., being taken seriously).[15] This is important given the economic toll pediatric chronic pain has on families and society.[13] The impact of pain education on diagnostic uncertainty in youth and their parents may be a particularly fruitful area for future research.[31]

Diagnostic uncertainty seemed to vary in intensity across participants. While some individuals expressed acceptance of the diagnosis, they continued to pursue other clinicians and additional diagnostic tests in a quest for answers and to exhaust other possible explanations. Parents found themselves caught between two opposing notions of being a good parent (i.e., protecting their child from potential harm of invasive procedures vs. not ending the search until the pain was 'fixed'), which was distressing. This is consistent with previous literature on parents' experience of anguish when they are not able to 'make things better' for their child with pain.[16] Enhanced understanding about how parental diagnostic uncertainty influences children's uncertainty, adjustment, and adherence to treatment, and pain outcomes, is important for future research. This qualitative analysis demonstrated that parents and youth are not always in agreement in terms of their pain conceptualization and 'buy-in'. What this lack of concordance means for children's outcomes is currently unknown.

How pain is conceptualized has been shown to be important for pain knowledge and catastrophizing, although its impact on disability is unclear.[11] Moreover, how a diagnosis of chronic pain is communicated to youth and families is likely critical to their 'buy-in,' and is likely influenced by developmental factors and individual needs. A better understanding of diagnostic uncertainty, between and within parent-youth dyads, may help tailor how clinicians deliver diagnoses to families to achieve 'buy in', increase understanding of pain and diagnosis, and ultimately improve treatment engagement and response. Youth also reported sensing uncertainty in clinicians providing the diagnosis and misalignment in agendas between themselves and clinicians. Frustration has previously been reported by children with chronic pain during medical encounters when they feel they have something to contribute but their narratives and beliefs are not elicited.[4] In the current sample, misalignment in patient-clinician communication and expectations was tied to diagnostic uncertainty and (mis)trust in the medical system. However, alignment between the orientation of patients (and caregivers) and their clinicians (e.g., if the patient is uncertain *and* perceives that the clinician is also uncertain about the pain) may also play a role in acceptance of a diagnosis and fuel diagnostic uncertainty. Past experiences of medical encounters and a learning history of incorrect diagnoses from clinicians fuelled uncertainty, demonstrating the importance of memories of previous medical care in influencing the experience of diagnostic uncertainty.

This research should be interpreted in light of limitations. The current data are from the perspectives of youth and parents and thus, the information families were provided with regards to the youth's diagnosis is unknown. Future research should investigate how aspects of the clinician-patient encounter (e.g., type of clinician, provision of pain education) when a diagnosis is provided, influence parents' and youth's memories of the encounter, their perceptions of the

diagnosis, and the impact of communication on diagnostic uncertainty. In addition, interviews were conducted at a single time point; thus, this study provided a snapshot of parents and youth at a particular time in their pain journey. It is likely that perceptions of diagnostic uncertainty are dynamic over time and are influenced by experiences and developmental factors. Future research might consider contacting families following the provision of a diagnosis to explore their interactions with clinicians and identify potential ways to reduce uncertainty. Finally, parent data in this study largely reflect maternal accounts of diagnostic uncertainty due to the low number of fathers (n=3) who participated. Furthermore, participants in the current sample predominantly reported high socioeconomic status, thus these accounts may not be representative of families from lower socioeconomic status who may face additional challenges (e.g., stigma, lack of resources) affecting diagnostic uncertainty.

This study highlights diagnostic uncertainty as a core feature of pediatric chronic pain. Given that untreated chronic pain in childhood can lead to persistent pain problems[42] and internalizing mental health disorders[27] into adulthood, investigation of diagnostic uncertainty and its impact on treatment engagement and response in this population is warranted. Illness uncertainty in pediatric chronic illness has been linked to worse psychological functioning and illness-related distress.[38] Among adults with chronic pain, diagnostic uncertainty is associated with poorer pain outcomes.[12,33] Further research is needed to understand factors that influence diagnostic uncertainty in youth and their parents, as well as the impact of diagnostic uncertainty on chronic pain and treatment outcomes over time. Research suggests that over 40% of adult patients with chronic pain[32] and parents of youth with chronic pain experience diagnostic uncertainty.[26] Our data suggest this is also frequently experienced by youth themselves. Nevertheless, the prevalence of diagnostic uncertainty in pediatric chronic pain populations

should be established. Pincus and colleagues[30] provide suggestions on how diagnostic uncertainty can be quantified in future research. This study suggests that there may be differences in how diagnostic uncertainty is perceived by parents and youth. Given that parental beliefs can impact children's medically unexplained symptoms,[9] parents' perceived diagnostic uncertainty warrants further investigation. If parent and/or youth diagnostic uncertainty is associated with response to pain treatment, interventions to decrease diagnostic uncertainty could have far-reaching implications across pain outcomes and health care expenditures.

References

1. Bhise V, Rajan SS, Sittig DF, Morgan RO, Chaudhary P, Singh H. Defining and Measuring Diagnostic Uncertainty in Medicine: A Systematic Review. *J Gen Intern Med.* 33:103-115, 2018

2. Braun V, Clarke V. Using thematic analysis in psychology. *Qualitative Research in Psychology* 3:77-101, 2006

3. Brodwall A, Glavin K, Lagerlov P. Parents' experience when their child has chronic abdominal pain: a qualitative study in Norway. *BMJ Open.* 8:e021066, 2018

4. Carter B. Chronic pain in childhood and the medical encounter: Professional ventriloquism and hidden voices. *Qual Health Res.* 12:28-41, 2002

5. Eccleston C, Crombez G, Scotford A, Clinch J, Connell H. Adolescent chronic pain: patterns and predictors of emotional distress in adolescents with chronic pain and their parents. *Pain.* 108:221-229, 2004

6. Eccleston C, Malleson P. Managing chronic pain in children and adolescents. We need to address the embarrassing lack of data for this common problem. *BMJ.* 326:1408-1409, 2003

7. Edmond SN, Keefe FJ. Validating pain communication: current state of the science. *Pain.* 156:215-219, 2015

8. Elliott R, Fischer CT, Rennie DL. Evolving guidelines for publication of qualitative research studies in psychology and related fields. *Br J Clin Psychol.* 38 (Pt 3):215-229, 1999

9. Eminson DM. Medically unexplained symptoms in children and adolescents. *Clinical Psychology Review.* 27:855-871, 2007

10. Friedrichsdorf SJ, Giordano J, Desai Dakoji K, Warmuth A, Daughtry C, Schulz CA. Chronic Pain in Children and Adolescents: Diagnosis and Treatment of Primary Pain Disorders in Head, Abdomen, Muscles and Joints. *Children (Basel).* 3, 2016

11. Gallagher L, McAuley J, Moseley GL. A randomized-controlled trial of using a book of metaphors to reconceptualize pain and decrease catastrophizing in people with chronic pain. *Clin J Pain.* 29:20-25, 2013

12. Geisser ME, Roth RS. Knowledge of and agreement with chronic pain diagnosis: Relation to affective distress, pain beliefs and coping, pain intensity, and disability. *J Occup Rehabil.* 8:73-88, 1998

13. Groenewald CB, Essner BS, Wright D, Fesinmeyer MD, Palermo TM. The economic costs of chronic pain among a cohort of treatment-seeking adolescents in the United States. *J Pain.* 15:925-933, 2014

14. Harris PA, Taylor R, Thielke R, Payne J, Gonzalez N, Conde JG. Research electronic data capture (REDCap)--a metadata-driven methodology and workflow process for providing translational research informatics support. *J Biomed Inform.* 42:377-381, 2009

15. Holt N, Pincus T, Vogel S. Reassurance during low back pain consultations with GPs: a qualitative study. *Brit J Gen Pract.* 65:E692-E701, 2015

16. Jordan AL, Eccleston C, Osborn M. Being a parent of the adolescent with complex chronic pain: An interpretative phenomenological analysis. *European Journal of Pain.* 11:49-56, 2007

17. King S, Chambers CT, Huguet A, MacNevin RC, McGrath PJ, Parker L, MacDonald AJ. The epidemiology of chronic pain in children and adolescents revisited: a systematic review. *Pain.* 152:2729-2738, 2011
18. Kool MB, van Middendorp H, Boeije HR, Geenen R. Understanding the Lack of Understanding: Invalidation From the Perspective of the Patient With Fibromyalgia. *Arthrit Rheum-Arthr.* 61:1650-1656, 2009
19. Linton SJ, Boersma K, Vangronsveld K, Fruzzetti A. Painfully reassuring? The effects of validation on emotions and adherence in a pain test. *Eur J Pain.* 16:592-599, 2012
20. Lynch-Jordan AM, Sil S, Peugh J, Cunningham N, Kashikar-Zuck S, Goldschneider KR. Differential changes in functional disability and pain intensity over the course of psychological treatment for children with chronic pain. *Pain.* 155:1955-1961, 2014
21. Mason J: Semi structured interview. In: Encyclopedia of Social Science Research Methods.(Lewis-Beck, M., Bryman, A, Liao, TF., Ed.), SAGE, Thousand Oaks, CA, 2004.
22. Meldrum ML, Tsao JC, Zeltzer LK. "I can't be what I want to be": children's narratives of chronic pain experiences and treatment outcomes. *Pain Med.* 10:1018-1034, 2009
23. Merskey H, Bogduk, N.: Classification of chronic pain: Descriptions of chronic pain syndromes and definition of pain terms. Report by the International Association for the Study of Pain Task Force on Taxonomy. . 2nd edition, IASP Press, Seattle, 1994.
24. Morrow SL. Quality and trustworthiness in qualitative research in counseling psychology. *J Couns Psychol.* 52:250-260, 2005
25. Moulin V, Akre C, Rodondi PY, Ambresin AE, Suris JC. A qualitative study of adolescents with medically unexplained symptoms and their parents. Part 2: How is healthcare perceived? *J Adolesc.* 45:317-326, 2015
26. Noel M, Beals-Erickson SE, Law EF, Alberts NM, Palermo TM. Characterizing the Pain Narratives of Parents of Youth With Chronic Pain. *Clin J Pain.* 32:849-858, 2016
27. Noel M, Groenewald CB, Beals-Erickson SE, Gebert JT, Palermo TM. Chronic pain in adolescence and internalizing mental health disorders: a nationally representative study. *Pain.* 157:1333-1338, 2016
28. O'Reilly M, Parker N. 'Unsatisfactory saturation': A critical exploration of the notion of saturated sample sizes in qualitative research. *Qualitative Research.* 13:190-197, 2012
29. Palermo TM. Impact of recurrent and chronic pain on child and family daily functioning: a critical review of the literature. *J Dev Behav Pediatr.* 21:58-69, 2000
30. Pincus T, Noel M, Jordan A, Serbic D. Perceived diagnostic uncertainty in pediatric chronic pain. *Pain.* 2018
31. Robins H, Perron V, Heathcote LC, Simons LE. Pain Neuroscience Education: State of the Art and Application in Pediatrics. *Children-Basel.* 3, 2016
32. Serbic D, Pincus T. Diagnostic uncertainty and recall bias in chronic low back pain. *Pain.* 155:1540-1546, 2014
33. Serbic D, Pincus T, Fife-Schaw C, Dawson H. Diagnostic uncertainty, guilt, mood, and disability in back pain. *Health Psychol.* 35:50-59, 2016
34. Shenton AK. Strategies for ensuring trustworthiness in qualitative research projects. *Education for Information.* 22:63-75, 2004
35. Sibeoni J, Chambon L, Pommepuy N, Rappaport C, Revah-Levy A. Psychiatric care of children with autism spectrum disorder - What do their siblings think about it? A qualitative study. *Autism.*1362361317728435, 2017

36. Sibeoni J, Orri M, Podlipski MA, Labey M, Campredon S, Gerardin P, Revah-Levy A. The Experience of Psychiatric Care of Adolescents with Anxiety-based School Refusal and of their Parents: A Qualitative Study. *J Can Acad Child Adolesc Psychiatry.* 27:39-49, 2018
37. Spencer L, Ritchie J, Lewis J, Dillon L: Quality in qualitative evaluation: A framework for assessing research evidence, National Centre for Social Research 2003.
38. Szulczewski L, Mullins LL, Bidwell SL, Eddington AR, Pai ALH. Meta-Analysis: Caregiver and Youth Uncertainty in Pediatric Chronic Illness. *J Pediatr Psychol.* 42:395-421, 2017
39. Tong A, Sainsbury P, Craig J. Consolidated criteria for reporting qualitative research (COREQ): a 32-item checklist for interviews and focus groups. *Int J Qual Health Care.* 19:349-357, 2007
40. van der Weijden T, van Bokhoven MA, Dinant GJ, van Hasselt CM, Grol RPTM. Understanding laboratory testing in diagnostic uncertainty: a qualitative study in general practice. *Brit J Gen Pract.* 52:974-980, 2002
41. van Ravesteijn H, van Dijk I, Darmon D, van de Laar F, Lucassen P, Hartman TO, van Weel C, Speckens A. The reassuring value of diagnostic tests: a systematic review. *Patient Educ Couns.* 86:3-8, 2012
42. Walker LS, Sherman AL, Bruehl S, Garber J, Smith CA. Functional abdominal pain patient subtypes in childhood predict functional gastrointestinal disorders with chronic pain and psychiatric comorbidities in adolescence and adulthood. *Pain.* 153:1798-1806, 2012

Table 1. Sociodemographic Characteristics of the Sample

	N	Mean	Range
Youth's age, years	20	14.6	10-18
Parent's age, years	18[a]	46.8	33-55
Relationship to youth			
Biological Mother	17		
Biological Father	3		
Youth's ethnicity (%)			
White (Caucasian)	18		
Latin American	1		
Do not want to answer	1		
Household income			
<$10,000 to $29,999	1		
$30,000 to $59,999	1		
$60,000 to $89,999	1		
More than $90,000	15		
Do not want to answer	2		
Marital Status			
Married	14		
Single	1		
Separated/divorced	3		
Widowed	1		
Common-law	1		

[a]Data only reported on 18 parents as two parents did not report their age

Table 2. Semi-structured Interview Schedule

Interview Questions
1. Where did this all start?
2. Where are you now in your journey?
3. Where do you see this going?
4. What do you remember about getting the diagnosis and what do you make of that?